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Risk factors for stuttering: a secondary analysis of a large data base

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Abstract The spectrum of risk and concomitant factors in stuttering is generally thought to be wide and heterogeneous. However, only a few studies have examined these factors using information from large databases. We examined the data on 11,905 Swiss conscripts from 2003. All cases with high psychiatric screening scores indicating “caseness” for a psychiatric disorder were excluded, among them potential malingers, so that 9,814 records remained. The analyses rely on self-reported information about stuttering in childhood, problems at birth, problems in school, mental disorders of parents and relatives, childhood adversity and socio-demographic information. Statistical modelling was done using logistic regression and path analysis models. Risk factors determined in the logistic regression include premature birth, probable attention deficit hyperactive disorder, alcohol abuse of the parents, obsessive–compulsive disorder in parents and relatives, having a disabled mother and having a parent from a foreign country. There is no overwhelmingly strong risk

factor; all odds ratios are about 2 or below. In conclusion, large databases are helpful in revealing less obvious and less frequent risk factors for heterogeneous disorders such as stuttering. Obviously, not only secondary analyses, but also systematical large scale studies would be required to complete the complex epidemiological puzzle in stuttering. An extensive examination of young adults who were initially assessed in childhood might provide the most promising design.

Keywords Stuttering · Epidemiology · Risk factors

Introduction

Stuttering is a relatively common disorder of verbal fluency that emerges in childhood in about 5% of children [16]. It is typically manifested at the beginning of long words and complex sentences. The disorder may have serious social and psychological sequels for the person so affected, especially if it persists beyond adolescence, which is the case in one out of five persons with stuttering [2, 32]. The empirical findings have thus far suggested some basic differentiations [2, 4, 7, 16]. These include (1) distinguishing developmental, or idiopathic stuttering, from acquired stuttering, (2) distinguishing persistent stuttering from recovering stuttering, which occurs in about four-fifths of all cases, (3) distinguishing stuttering in the presence of family history of stuttering from stuttering where there is no family history; and finally, (4) distinguishing stuttering with comorbidity with other disorders from stuttering without comorbid conditions.

Since various brain areas and tracts are involved in the planning and producing of fluent speech [7, 15, 23, 32], a wide spectrum of etiological patterns and associated risk factors, correlates and intervention approaches, may be

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assumed. A wide range of possible risk factors has been proposed in literature including male sex, low intelligence scores, prenatal, perinatal or other brain damage, but also neurological lesions (particularly in stuttering without hereditary evidence), concussion and head injury, genetic risk, delayed speech development, attention deficit hyperactive disorder (ADHD) and associated symptoms (e.g. enhanced gross motor activity, impulsivity, decreased attentional focusing, decreased inhibitory control, perceptual sensitivity), developmental coordination disorder, learning disorders, emotionally reactive and sensitive temperament, neurotic conflicts and impaired child–parent interactions, negative parental reactions to normal childhood disfluency [2–4, 10, 16, 26].

It may reasonably be assumed that this list is far from complete. Till now, research into stuttering has only rarely made use of large epidemiological databases [14, 29] and challenging statistical models [21], which might provide valuable information on rare or less obvious risk factors. This paper aims to fill this gap by presenting secondary analyses carried out on a database of about 10,000 Swiss conscripts.

Methods

Sample

The sample comprises conscripts who attended recruitment to the Swiss Armed Forces in 2003. The Swiss Armed Forces introduced an all-new recruitment procedure for conscripts and female volunteers in stages during that year. A group of 14,157 individuals (out of 24,292 conscripts and 123 volunteers) underwent the new recruitment procedure, including a psychiatric screening that used a comprehensive questionnaire as its first step. After the exclusion of incomplete or erroneous records there were 12,523 records remaining among them 11,905 male conscripts aged 18–20. In the next step, all cases with high psychiatric screening scores were excluded, among them most potential malingerers who generally represent a major problem in conscription screening. The criteria for exclusion relied on the Symptom Checklist-90-Revised (SCL-90-R) caseness definition [13], and entailed either the Global Severity Index (GSI) score ≥ 63 (after T-standardization) or two or more subscale scores ≥ 63 (after T-standardization). Thus, further 2,091 data records were excluded from the analysis, whereas 9,814 records remained.

Questionnaires

The conscription procedure comprises, among other elements, a psychiatric screening. All screening sessions are introduced and supervised by military test psychologists.

The screening includes the SCL-90-R [12] as an externally validated psychometric instrument. Further questionnaires assess information on psychopathological symptoms, on substance use and other behavioural characteristics of the conscripts, on mental and school problems in childhood and youth, on childhood adversity issues, on mental disorders of parents and relatives, and additional demographic information. Regarding former mental and school problems the questionnaires assess stuttering (“Did you stutter in childhood?”), probable dyslexia (“Do you have problems with reading and writing?”), attention deficit hyperactive disorder (“Were you restless or fidgety at school?”) and several other issues. Additional questions assess problems at birth (“Were there any complications at your birth?”), and spending the first days of life in an incubator.

All information relies on self-reporting by the conscripts. The use of anonymised information in epidemiologic studies was cleared by the Zurich State Ethical Committee (KEK) to fulfil all legal and data privacy protection exigencies.

Statistical modelling

For the preliminary bivariate analyses, we classified the putative correlates of stuttering either as putative risk factors (or putative indicators of risk factors), concomitant factors, and variables not unequivocally assignable as either risk or concomitant factor. In the subsequent multivariate analysis, i.e. logistic regression, we included only variables assigned as putative risk factors. Finally, we differentiated background (parental, family) risk factors, and intermediate risk factors in a path analysis model. The path analysis was based on logistic regressions for each intermediate risk factor. Irrelevant risk factors were excluded from the final model.

We analysed the data with common statistical models such as correlational and logistic regression analysis. Data compilation and initial analyses were performed using SAS for Windows (Version 8). The path analysis was performed with Mplus (Version 4.21).

Results

In the restricted sample, 408 men (4.2% out of 9,814) reported having stuttered in childhood. Without the elimination of malingerers and other SCL-90-R-high-scorers, this group would have contained 601 men (5.1% out of 11,905).

Table 1 shows the results from the bivariate analyses. The upper part of the table represents putative risk factors (or putative indicators of risk factors), and the lower part the concomitant factors. In between are variables which are not unequivocally assignable to either risk or concomitant

Table 1 Bivariate analyses of risk and concomitant factors of persisting or recovered stuttering in Swiss male conscripts 2003

Variable	Overall <i>N</i>	Stuttering <i>N</i>	OR	Confidence interval
Putative risk factors				
Troubles at birth	1,091	65	1.55	1.17–2.03
Incubator	1,012	61	1.56	1.18–2.06
Restless and fidgety in school	1,831	105	1.54	1.22–1.93
Mother alcoholic	88	10	3.00	1.54–5.84
Father alcoholic	305	25	2.13	1.39–3.24
Mother with other addiction	202	8	0.95	0.46–1.93
Father with other addiction	299	18	1.50	0.92–2.43
Anxiety in family members/relatives	324	21	1.63	1.03–2.56
Compulsion in family members/relatives	254	19	1.91	1.18–3.07
Depression in family members/relatives	1,067	44	0.99	0.71–1.36
Schizophrenia in family members/relatives	605	23	0.91	0.58–1.39
Family member was psychiatric inpatient	554	28	1.24	0.83–1.84
Mother is disabled ^a	114	12	2.76	1.50–5.06
Father is disabled ^a	194	14	1.82	1.04–3.16
Mother had a serious physical disease	262	19	1.84	1.14–2.96
Father had a serious physical disease	307	20	1.64	1.02–2.60
Parent from a foreign country	1,842	98	1.39	1.09–1.75
Risk or concomitant childhood factors				
Sibling is disabled ^a	188	6	0.76	0.33–1.71
Sibling had a serious physical disease	119	7	1.45	0.67–3.12
Frequently beaten by parents or educators	225	10	1.07	0.56–2.03
Not grown up with both parents	1,911	91	1.19	0.94–1.51
Raised (at least for a while) in an asylum	267	22	2.13	1.36–3.33
Raised (at least for a while) with grandparents	806	47	1.48	1.08–2.02
Hospitalised because of head injury	1,162	65	1.43	1.09–1.88
Concomitant childhood factors				
Came late to school (8 years or older)	114	18	4.47	2.67–7.47
Examined by school psychologist	755	50	1.72	1.26–2.33
Troubles with reading and writing	474	57	3.50	2.59–4.70
Teased for deformity or did not come to terms with deformity	750	52	1.82	1.34–2.46
Had psychological counselling and/or therapy	749	45	1.53	1.11–2.10

Baseline *N*: 9,814, persons with stuttering: 408, relevant odds ratios are shown in bold letters

^a Since childhood or after an accident

factors. The results suggest that only small proportions of persons with stuttering are influenced by each putative risk or concomitant factor.

At first glance, several groups of risk factors can be differentiated:

- Birth-related problems, as indicated in approximately 10% of the conscripts, and 15% persons with a stuttering problem. The “incubator” item assigns premature births as a risk factor for stuttering. The odds ratios for both risk factors have a similar magnitude (~ 1.5).
- Having been restless and fidgety at school points to a probable ADHD background. Such behaviour was

reported by one in four conscripts who reported also a stuttering condition

- Among the substance variables, both parents’ alcohol variables yielded relevant odds ratios between 2 and 3, but not for the variables related to “other substance” disorders.
- Anxiety disorders and obsessive–compulsive disorders (OCD) of family members and other relatives had a noteworthy predictive effect, but no other disorders such as depression or schizophrenia. Also the “inpatient” item, which is a proxy for serious psychiatric disorders in family members and relatives, proved to be not relevant.

- Disabling disorders and serious physical diseases of the parents yielded noteworthy odds ratios. The odds ratio of the item “disabled mother” is nearly as high as the odds ratio regarding mother’s alcohol addiction.
- Having a parent from a foreign country slightly enhanced the risk of stuttering.
- Being raised separately from the parents includes items which need further analyses and information, because the temporal sequence of the effects is not obvious.

The concomitant variables (came late to school, saw the school psychologist, had difficulty with reading and writing, was teased for deformity, underwent psychological counselling and/or therapy) correspond to delayed intellectual development and to the psychosocial consequences of stuttering. Difficulty with reading and writing is a putative indicator for dyslexia, but possibly also secondary analphabetism.

In multivariate logistic regression analysis we included the risk factors and, in addition, the “frequently beaten” variable since we assumed that violence against the child typically had begun in its first years of life (Table 2). The number of relevant predictors decreased in multivariate analysis, however, the spectrum remained as broad as before. The variables that remained relevant after adjustment for the other factors are: incubator (i.e. premature birth), having been restless and fidgety at school (probable ADHD), alcohol abuse of the parents, OCD in family members and relatives, and having a parent from a foreign country.

The path analysis (Fig. 1; Table 3) helped to differentiate the intermediate risk factors from the background

parental variables. Only a few background variables act simultaneously on stuttering and on any related intermediate variable. Alcohol abuse of the mother acts over three pathways: a direct pathway, an indirect pathway involving the incubator (premature birth) variable, and another indirect pathway involving having been “restless and fidgety at school” (probable ADHD). Since the latter variables are not associated, one may assume at least two different biological mechanisms representing the effects of alcohol abuse. The effects are similar to having a “parent from a foreign country” whose effect may be direct or mediated by the subject having been “restless and fidgety at school” (probable ADHD) or mediated by the more complex path leading over “frequently beaten” and “restless and fidgety at school” (probable ADHD). However, the fact that “frequently beaten” and stuttering are not interrelated clearly indicates two different mechanisms.

Moreover, the path analysis adds some perspectives on the risk factors themselves. Alcohol and substance abuse of the mother seem to be the most multifarious risk factor, but also “schizophrenia in family members/relatives” and having a “parent from a foreign country” are prominent.

Discussion

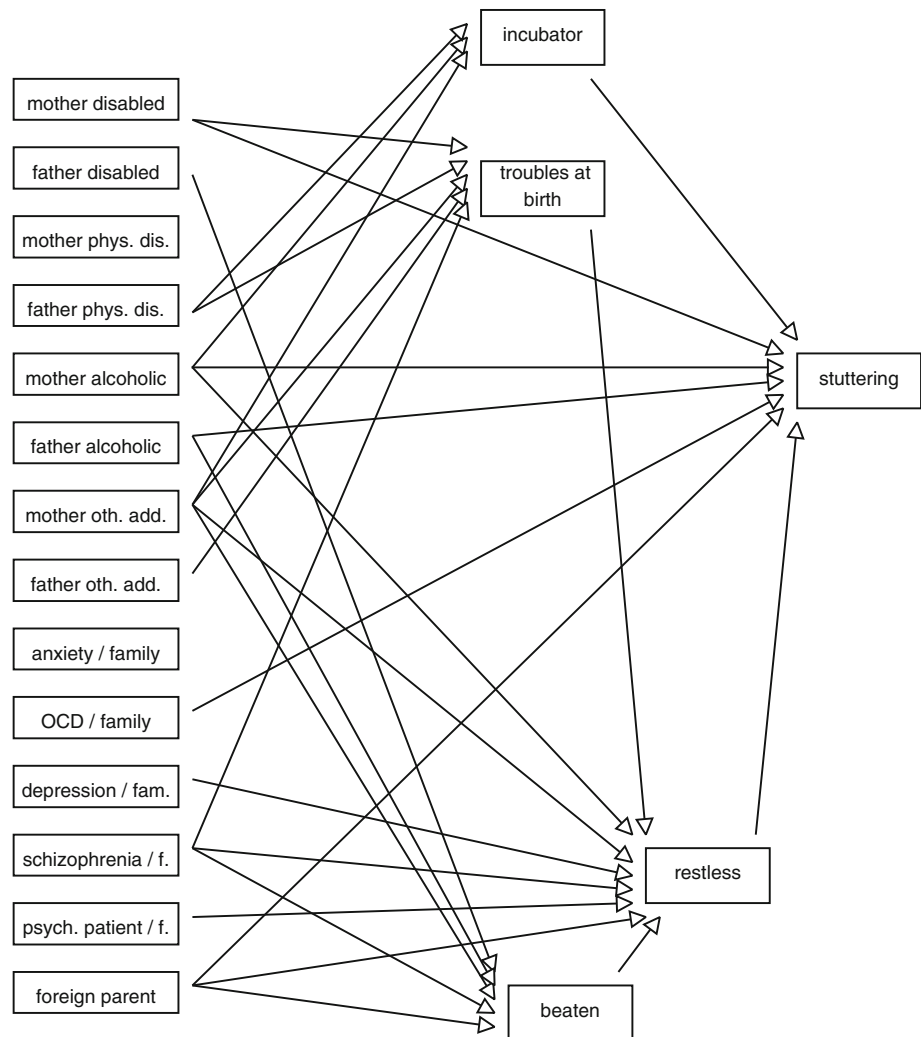
This study explored the spectrum of risk and concomitant factors of stuttering using data from a large population database. The data were derived from the psychiatric and psychological screening of Swiss conscripts in 2003, and

Table 2 Logistic regression analysis: risk factors of persisting or recovered stuttering in Swiss male conscripts 2003

Variable	Estimates	SE	OR	Confidence interval
Troubles at birth	0.25	0.15	1.28	0.95–1.72
Incubator	0.35	0.15	1.41	1.04–1.90
Restless and fidgety in school	0.36	0.11	1.44	1.14–1.81
Mother alcoholic	0.77	0.37	2.16	1.04–4.46
Father alcoholic	0.63	0.22	1.88	1.20–2.95
Mother with other addiction	−0.65	0.42	0.52	0.22–1.20
Father with other addiction	0.28	0.27	1.32	0.76–2.29
Anxiety in family members/relatives	0.42	0.24	1.52	0.93–2.47
Compulsion in family members/relatives	0.60	0.25	1.82	1.10–3.01
Depression in family members/relatives	−0.19	0.17	0.82	0.58–1.16
Schizophrenia in family members/relatives	−0.35	0.23	0.70	0.44–1.11
Family member was psychiatric inpatient	0.11	0.21	1.11	0.72–1.70
Mother is disabled	0.66	0.34	1.93	0.98–3.78
Father is disabled	0.29	0.31	1.33	0.72–2.47
Mother had a serious physical disease	0.42	0.26	1.53	0.91–2.55
Father had a serious physical disease	0.24	0.25	1.28	0.76–2.11
Parent from a foreign country	0.29	0.12	1.34	1.05–1.69
Frequently beaten by parents or educators	−0.33	0.35	0.72	0.36–1.43

Relevant odds ratios are shown in bold letters

Fig. 1 Path analysis of background and intermediate risk factors of persisting or recovered stuttering in Swiss male conscripts 2003 (see also Table 3)



covered a broad spectrum of psychological items and psychiatric symptoms, among them rare risk and concomitant factors. We could identify a set of well-known correlates, as well as rare or new putative risk factors. Among the latter were premature birth, alcohol abuse of the parents, and having a parent from a foreign country.

Methodological considerations

As is common in secondary analyses of existing epidemiological data, this is a hypothesis-generating study. However, the context of mandatory military conscription delivers data, which may be quite different from the common context of a representative population-based study. Thus, we omitted all cases with high psychiatric symptom loads in order to avoid systematic bias that we might expect with regards to malingering. This equally resulted in omitting an unknown number of conscripts with valid responses, but high comorbidity. Provided that the major sources of systematic biasing effects (malingering,

comorbidity) were excluded, the remaining source of bias, for example, self-reporting, are few. More importantly, they mainly yield noise in the data, and, as a consequence, smoothed instead of biased results. We have to expect that some of the effects that we found in this study are stronger in reality, and that we possibly missed some weak effects due to noisy data. Since we are dealing with risk factors, the consequences are not as grave as in studies focusing mainly on prevalence or incidence estimates.

Interpretation of the results

In accordance with the assumption that stuttering is a heterogeneous disorder, we found risk factors that obviously represented a wide spectrum of etiological mechanisms and related disturbances. There is no one overwhelming strong risk factor; all odds ratios are about 2 or below.

Having been “restless and fidgety in school” was an expected correlate of stuttering representing ADHD [2, 6].

Table 3 Path analysis of background and intermediate risk factors of persisting or recovered stuttering in Swiss male conscripts 2003

Variable	Estimates	SE	OR	Confidence interval
Incubator ON				
Mother alcoholic	0.62	0.28	1.86	1.08–3.20
Mother with other addiction	0.49	0.19	1.63	1.12–2.39
Father had a serious physical disease	0.37	0.17	1.45	1.04–2.00
Troubles at birth ON				
Father had a serious physical disease	0.54	0.15	1.72	1.28–2.32
Father with other addiction	0.48	0.16	1.62	1.17–2.23
Mother with other addiction	0.54	0.19	1.71	1.18–2.49
Schizophrenia in family members/relatives	0.32	0.12	1.38	1.09–1.75
Mother is disabled	0.60	0.24	1.83	1.14–2.93
Frequently beaten by parents or educators ON				
Father is disabled	0.74	0.34	2.09	1.07–4.08
Father alcoholic	1.60	0.21	4.95	3.29–7.44
mother with other addiction	1.30	0.27	3.68	2.18–6.20
Schizophrenia in family members/relatives	0.56	0.22	1.75	1.15–2.67
Parent from a foreign country	0.91	0.14	2.49	1.88–3.30
Restless and fidgety in school ON				
Troubles at birth	0.35	0.08	1.42	1.22–1.65
Mother alcoholic	0.93	0.23	2.55	1.64–3.97
Mother with other addiction	0.51	0.16	1.66	1.22–2.27
Depression in family members/relatives	0.28	0.08	1.32	1.12–1.55
Schizophrenia in family members/relatives	0.35	0.10	1.42	1.17–1.72
Family member was psychiatric inpatient	0.29	0.11	1.34	1.08–1.65
Parent from a foreign country	0.30	0.06	1.35	1.19–1.53
Frequently beaten by parents or educators	1.02	0.14	2.77	2.10–3.65
Stuttering ON				
Incubator	0.42	0.14	1.53	1.15–2.03
Mother is disabled	0.81	0.32	2.26	1.20–4.24
Mother alcoholic	0.73	0.36	2.07	1.02–4.20
Father alcoholic	0.61	0.22	1.84	1.19–2.85
Compulsion in family members/relatives	0.56	0.25	1.75	1.08–2.86
Parent from a foreign country	0.29	0.12	1.34	1.06–1.69
Restless and fidgety in school	0.36	0.12	1.43	1.14–1.81

ADHD has been suggested to be associated with other related disorders such as dyslexia [25]. The underlying common etiologies may be both of hereditary nature as well associated with acquired or secondary disorders [2].

OCD in family members and relatives and other parental psychological influences were a favourite topic of psychological stuttering research in the twentieth century, but do not remain so [4]. It is remarkable that no other psychiatric disorder of parents occurs in the list of risk factors in our large database. Thus, one might speculate that some pathways in OCD and in stuttering share a genetic and biological background. For example, basal ganglia circuits have been suggested as a possible link between stuttering and OCD [1], but also may be involved in ADHD [28].

Some relevant items found in our analysis have been infrequently reported. Premature birth has occasionally

been presented as a risk factor for stuttering [2, 18, 20]. In contrast to other studies [8, 27], no effect on probable ADHD could be shown in the Swiss conscripts data. The missing link might be due to the fact that premature birth appears to be associated with attention problems rather than with hyperactivity problems [19]. Interestingly, the item “complications at birth” turned out to be relevant as regards probable ADHD but not as regards stuttering behaviour in multivariate analysis. Probably, the results were shaped by the fact that many participants equated premature births with problems at birth (about 40% redundant positive answers in each variable).

Alcohol abuse of the parents also needs further examination. Alcohol abuse has been rarely documented as a risk factor in stuttering [22]. Alcohol, but not other substances, seems to have a crucial effect on stuttering in our analysis.

This indicates that the association is hardly related to adverse social environments, but rather to the foetal alcohol spectrum disorders (FASD). As we have no exact information about the parents' age of alcohol abuse, we may assume that misuse began during adolescence and young adulthood, which is typical of the initiation of substance misuse patterns. Alcohol misuse by the pregnant mother is known to have serious effects on the prenatal brain development leading to FASD [30, 31]. FASD are known to be associated with learning disabilities, lower IQ, ADHD, and motor impairment. Similar, eventually less severe consequences may also result from occasional alcohol misuse [9]. In contrast to the mother's alcohol abuse, alcoholism of the father as a risk factor for stuttering behaviour is more difficult to interpret. Possibly, this variable is a proxy for the alcohol misuse of the mother at lower exposure levels. An alternative explanation might suggest impulsiveness and aggressive behaviour by the alcoholic father as possible intermediate mechanisms. However, this is not supported by the variable "frequently beaten". As with other variables indicating adverse social environment, it proved not to be relevant with regards to stuttering.

Having a parent from a foreign country is an intriguing, although not a particularly strong, putative risk factor. The first hypothesis, impulsiveness and aggression deriving from the association with the variable "frequently beaten", was not supported by this analysis. Alternatively, "having a parent from a foreign country" might point at problems in language development in bilingual or multilingual families. A recent study has identified bilingual children to be at higher risk for stuttering than monolingual children or children speaking an alternative language exclusively [17].

"Having a disabled mother" is a risk factor which reappears in the list if a more narrow set of predictors was considered as is the case in the submodels within the path analysis. Since the path leading from "disabled mother" to "complications at birth" does not carry on to "stuttering", the focus should be on prenatal or postnatal factors. Unfortunately, no further information was available in our data which would have allowed us to be more specific.

Path analysis

The path analysis provides a more adequate perspective on the complex framework of background and intermediate variables involved in the prediction of a neurobehavioural symptom such as stuttering. Apart from risk factors generating direct effects on stuttering, there are also additional indirect paths. Comparing indirect paths may be helpful in revealing distinct mechanisms and processes leading to stuttering, even if they start with the same risk factor such as alcohol abuse of the mother.

Concomitant factors in stuttering

The associations with concomitant factors are in line with previous research. There is a close and well-known association between stuttering and dyslexia [5]. The odds ratios in this association, and with regard to delayed school start, are distinctly the strongest in our analysis. Having been teased for deformity and having had psychological counselling or treatment by the school psychologist or a psychotherapist are obvious psycho-social consequences of stuttering.

Limitations

This study relied on screening data, which were not specifically designed for the analysis of speech problems, such that various putatively interesting variables are missing in our analysis. In particular, it was not clear whether the results related to persistent stuttering or transient stuttering during childhood. More detailed information accessible in epidemiological surveys might include signs of stuttering [11], their persistence [5] and the differentiation from other speech disorders [24]. Moreover, no information was available in the Swiss conscripts data on stuttering in the family and among relatives. Some risk factors were represented by proxy variables such as premature birth ("incubator") and probable ADHD ("restless and fidgety in school"). The information relied on self-reporting of symptoms, problems and behaviour in a computer-assisted interview, and was therefore subject to recall bias. No clinical validation of any syndrome or any symptoms was available. The false-positive and false-negative self-reports have mostly a smoothing effect on the results.

Conclusion

Large databases are helpful in revealing some less obvious and less frequent risk factors for heterogeneous disorders, such as stuttering. Such factors found in this study include premature birth, alcohol abuse of the parents, or having a parent from a foreign country. Obviously, not only secondary analyses, but also systematical large scale studies, would be required to complete the complex epidemiological puzzle in stuttering. An extensive survey of young adults with stuttering and a control group, who were both initially assessed in childhood, might provide the most promising design. The survey should include detailed information on the neurological, psychiatric and other disabling disorders among the participants' parents and families.

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